

Uterine torsion in a 25-week pregnant female with congenital uterine didelphys and intraoperative complication of uterine atony: a report of a rare clinical case

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Uterine torsion is an extremely rare pregnancy-related complication. Its diagnosis is often difficult due to unspecific clinical symptoms, which can be mistaken for other conditions such as placental abruption. A 23-year-old pregnant woman with gestational age of 25 weeks presented with acute abdominal pain and hypertonic uterus. Vaginal examination revealed a septated vagina, double cervix, and double uterus. Bedside sonography revealed absence of fetal cardiac activity and signs of placental abruption. The patient was emergently operated with a preliminary diagnosis of severe placental abruption in the uterus didelphys. During surgery, we noted a double uterus and the fetus had implanted in the right uterus, which was twisted 180 degrees to the left. After detorsion, a low transverse hysterotomy was performed to extract the dead fetus. However, subsequently, the patient developed uterine atony. Subtotal hysterectomy and right adnexectomy were inevitable due to failure of conservative treatment with B-lynch suture. The patient's postoperative condition was stable, and she was discharged after 5 days.

Keywords

Uterine torsion; Pregnancy; Uterus didelphys; Uterine atony

1. Introduction

Uterine torsion is an extremely rare complication during pregnancy [1]. Medical literature has reported this as a "once-in-a-lifetime" diagnosis, and doctors usually encounter it once in their professional lives, at the most [2]. The exact incidence is currently unknown, whereas the rate of maternal and fetal mortality is significant at 12-18% [3, 4]. The clinical symptoms of this condition are non-specific, which makes the preoperative diagnosis very challenging [5]. This condition can cause numerous highly fatal complications such as placental abruption, stillbirth, uterine atony, and torsion of the urinary bladder [6]. Most cases are identified during emergency surgeries related to these complications. In this report, we present an emergency case with a 180-degree twisted uterus with uterus didelphys complicated by uterine atony in a pregnant woman in the 25th gestational week.

2. Case presentation

A 23-year-old primiparous woman, who was in the 25th gestational week, was admitted to the emergency unit of the Department of Obstetrics and Gynecology with symptoms of acute and sudden abdominal pain after an accidental fall. Her heart rate was 96 beats per minute, temperature was 37 degrees Celsius, blood pressure was 110/70 mm Hg, and respiratory rate was 20 per minute. Her cardiac examination was normal, except for mild systolic murmur over the aortic area. Other symptoms included severe abdominal pain, paleness, and sweating. Further clinical examination revealed continuous hypertonic uterine contraction, uterine tenderness, and abnormal abdominal shape. Uterine height and abdominal circumference were 26 and 83 cm respectively. Handheld fetal Doppler showed absence of fetal heart rate. Vaginal examination revealed a septated vagina, fully closed non-dilated double cervix, and double uterus of which the right uterus had a 6-month-old fetus with high fetal station. The patient had a reduced red blood cell count (2.8 M/L), hemoglobin level (8.5 g/dL), and hematocrit (25.6%). The results of coagulation tests (fibrinogen, thromboplastin time, blood circulation time, blood clotting time, prothrombin time) were within the normal limits. Emergency fetal ultrasound examination identified a fetus of 24 gestational weeks with no cardiac activity, placental abruption, and uterus didelphys. The patient was administered two units of red blood cells and was operated emergently with the following preliminary diagnosis: primiparous pregnancy in the 25th gestational week, severe placental abruption, and congenital uterine malformation (double uterus, double cervix, and double vagina).

Intraoperative finding showed that the right uterus, which had a 6-month-old fetus, was clockwise twisted in a semi-circle to the left along with the right adnexa, which was completely necrotic; the left uterus and adnexa were normal. After widening the abdominal incision and detorsion, a low transverse hysterotomy was performed, and a 600 g dead female fetus was extracted along with the fully separated placenta with amniotic fluid and blood clots. The right uterus

and adnexa were non-viable despite 15 minutes infusion of uterotonics and B-lynch suture (Fig 1,2). After counseling the patient's family about the fatal condition and failure of conservative treatment, subtotal hysterectomy and right adnexectomy were performed. After surgery, the complete blood count was repeated. The results of this test showed a hemoglobin level of 8.5 g/dL, and hematocrit of 25.7%. The patient was administered two more units of packed red blood cell, after which the hemoglobin level rose to 10.1 g/dL. Coagulation functions were within the normal range. The patient's postoperative condition was stable, and she was discharged after 5 days.

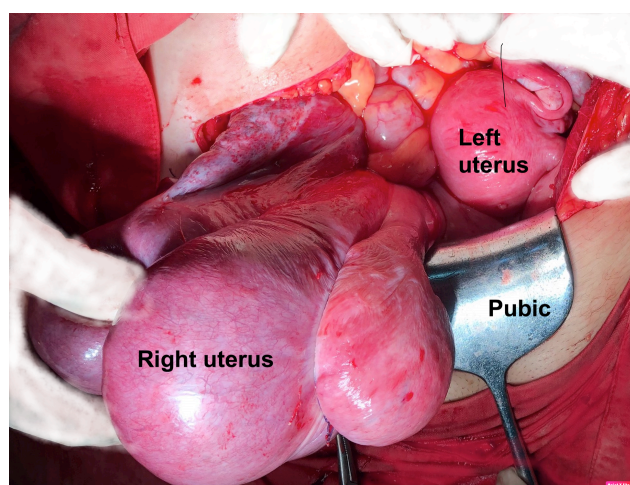


Fig. 1. The picture is taken from the right side and pointed from the patient's head. The uterus is left to normal and is on the right side of the picture. The uterus after untwisting, a lower horizontal incision was performed and a dead fetus was delivered, weighing 600 gr, a Lynch B stitched was sutured to stop bleeding due to uterine atony, the right appendages was wholly bruised.

This case-report was conducted with the patient's agreement and written informed consent was obtained. The study was conducted in accordance with the Declaration of Helsinki, and the protocol was approved by the Ethics Committee of [anonymous institution] (approval number H2020/384).

3. Discussion

Uterine torsion is defined as "the rotation of the uterus by more than 45 degrees around the long axis" and is rarely seen but it may occur among any gestational ages [4, 7, 8]. Torsion is usually by under 180 degrees, but cases of torsion ranging from 45 to 720 degrees have also been recorded in the medical literature [4]. In a study by author John Gronkjer Jensen, Denmark that included 212 cases of uterine torsion, 31% were less than 90 degrees; 58% ranged from 90 to 180 degrees, 6% ranged from 180 to 360 degrees, 3% were twisted by more than one full circle, and 2% had unspecified degrees of rotation [4].

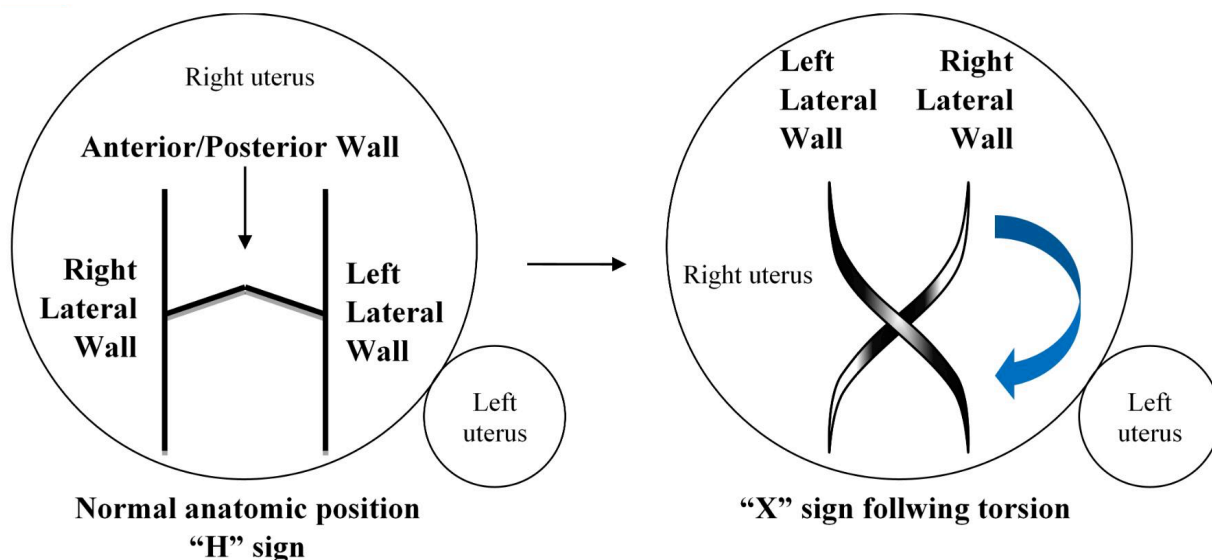
Almost all cases of uterine torsion described in the literature were associated with preexisting gynecologic conditions, such as uterine leiomyoma, congenital anomalies, adnexal mass, or fetal malpresentation [9]. Congenital uterine anomalies, which affect 1 in 201 women in the general population and are present in 1 in 594 fertile women, was one of the identified risk factors [10]. According to Jensen in a report on uterine torsion in 1976-1990, the number of cases of uterine abnormalities was 26, accounting for approximately 11%, mainly double or bicornuate uterus [4]. Fabien Demaria reported a case of a 23-year-old woman with twin pregnancy who was diagnosed with torsion of one horn of a gravid didelphic uterus during cesarean delivery after an erroneous diagnosis of abruptio placentae [11]. In 2012, Author Lucio M. A. Cipullo described a case of a uterine torsion in a woman carrying a Müllerian anomaly (uterus didelphys) [12], which is the known risk of hemi-uterus torsion, that occurred only during labor [13]. In those cases, the diagnosis was made during the cesarean section, performed in response to abnormal fetal tracing and failure of labor to progress.

Clinical symptoms of uterine torsion are typically diverse, with varying degrees and low specificity such as abdominal pain, vaginal bleeding, and dizziness, or are manifested as digestive or urinary symptoms due to compression [4, 14, 15]. However, approximately 11% of cases are asymptomatic. Therefore, clinical diagnosis of uterine torsion during pregnancy is very difficult. The pregnant patient might have abdominal pain due to uterine contraction, vaginal bleeding, delayed labor due to cervical complications, abnormal fetal presentation, and fetal distress, which can easily lead to a misdiagnosis.

Uterine twists to the right side are more common due to the physiological anatomy, as the uterus tends to skew to the right, especially during pregnancy [16]. However, one-third of uterine torsion cases still occur to the left, as seen in our case [17]. This can be explained by the fact that the fetus was implanted in the right uterus of a double uterus; therefore, the twisting could only occur to the left.

The exact cause of this complication remains unknown. It can occur in perfectly normal patients, irrespective of the age group, developmental stage, or pregnancy conditions. Nevertheless, many cases do occur in patients with uterine malformation, pelvic adhesions, abnormal fetal presentation, and hyperactive fetus, or injuries to the mother due to fall, traffic accidents, etc. [4]. Our patient had some risk factors that could have caused this complication, which includes double uterus and its occurrence after a fall. Our patient also had acute abdominal pain, and ultrasound revealed placental separation and dead fetus, which led to the diagnosis of severe placental abruption. The uterine torsion caused direct compression on the uterine and ovarian veins, which increased the pressure inside the placenta due to venous thrombosis, resulting in placental abruption and fetal death.

Diagnostic imaging including abdominal ultrasound, magnetic resonance imaging, and computed tomography can



Panel X.tif

Fig. 2. The normal anatomical position of the lower part of the vagina (H-shape) and the clockwise rotation of the right side uterus in uterine didelphys because the lower vagina is fixed at the introitus.

assist with the diagnosis. On magnetic resonance imaging (MRI) films, the X sign can be observed in the upper region of vagina or a narrow vagina in the upper region. Computed tomography can reveal torsion in the lower region of uterus and two ipsilateral adnexa. Some signs on ultrasound that can suggest uterine torsion are a change in the placental position, fetal tachycardia, absence of uterine blood flow on Doppler ultrasound, abnormal position of the ovary or signs of placental abruption, and fetal death [2, 18, 19].

In all cases, surgery is mandatory. Ideally, the uterus can be detorsioned to the normal position before uterine incision to extract the fetus. In difficult situations where the fetus is too large to untwist the uterus, a vertical incision is a safe option as the lower horizontal incision can cause injury to the uterine vessels and ovarian pedicles. Although uterine torsion is extremely rare, obstetricians should suspect this condition when patients have symptoms of acute abdominal pain and fetal distress, as seen in cases of placental abruption.

The degrees and duration of uterine torsion are essential factors contributing to the mortality rates of the fetus and the mother. The prognosis of mothers is usually good after undergoing surgery. However, the neonatal mortality rate is still at 12-18% and maternal mortality rate since 1976 has been 0% [4].

4. Conclusions

Although uterine torsion is very uncommon in real life and only intraoperative diagnosis is possible during emergency surgery, it is necessary to suspect uterine torsion in cases of acute abdominal pain and unexplained dizziness in pregnant patients with risk factors, especially congenital uterine malformation, pelvic mass, fetal malpresentation, or hyperactive fetus. A high level of suspicion and timely treat-

ment are important factors contributing to good prognosis for tackling uterine torsion.

Author contributions

CKT, TDT, NVQH, LMT designed and performed the research study. CKT and TDT collected data and taken the pictures. CKT, TDT and LMT wrote the manuscript. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

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Conflict of interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of this article.

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